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Incidental Liposarcomas Identified During Hernia Repair Operations

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Background and Objectives: Since the inguinal region communicates with the retroperitoneum, both retroperitoneal as well as de novo spermatic cord liposarcomas may be detected during hernia repair operations. We assessed the incidence of liposarcomas presenting at hernia repair in our hospital.
Methods: We performed a clerical review of pathology reports on adult tissue accessioned during hernia repair operations and reviewed operating room logs to obtain information concerning the total number of hernia repair operations (since some operations afford no accessioned tissue).
Results: Between 1992 and 1997, 1,736 adult hernia repair specimens were accessioned from approximately 2,000 operations. Among these, 22% had an associated cord lipoma; 2 cases were well-differentiated liposarcomas. These were from males aged 56 and 64 years in contrast to the mean age of 35 years for cord lipoma and measured 13 and 10 cm, whereas the mean size for cord lipomas was 5.5 cm. One of the liposarcomas had radiographic evidence of extension from a retroperitoneal lesion; the other appeared confined to the groin. On surgical exploration, the lesion was restricted to the spermatic cord region in both cases despite the suggestion of retroperitoneal extension/involvement in one.
Conclusions: Incidental liposarcomas identified during hernia operations are rare (<0.1% at our institution) but their presence merits histologic evaluation of adipose tissue from these cases. However, if efforts to contain costs are implemented and histologic review of such tissue is deemed generally unrewarding, large (>10 cm) fatty masses from this area should still be sampled. *J. Surg. Oncol. 1999:71:50–53.*

KEY WORDS: liposarcomas; inguinal hernias; dedifferentiation; soft-tissue tumors

INTRODUCTION

Since the inguinal region communicates with the retroperitoneum, both retroperitoneal as well as de novo spermatic cord liposarcomas may be detected during hernia repair operations. We assessed the incidence of liposarcomas presenting at hernia repair in our hospital.

MATERIALS AND METHODS

We performed a clerical review of pathology reports on adult tissue accessioned during hernia repair operations and reviewed operating room logs to obtain information concerning the total number of hernia repair operations (since some operations afforded no accessioned tissue). Pathology reports were reviewed to assess tumor size and basic demographic information and, in the two cases of liposarcomas, slides were reviewed and operative and follow-up information obtained. The clinical and surgical details of the two cases of inguinal liposarcoma were reviewed to identify clinical findings associated with this lesion.

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RESULTS

Between 1992 and 1997, 1,736 adult hernia repair specimens were accessioned for histology from 2,054 operations. Those in which no tissue was submitted were from direct hernias. Among the 1,736 specimens reviewed histologically, 382 (22%) had an associated cord lipoma; two cases among these were well-differentiated liposarcomas.

Clinical Characteristics of Inguinal Liposarcomas

The patients with inguinoscrotal liposarcomas were males aged 56 and 64 years. While males of any age may develop inguinal hernias, the mean age for all adult hernia patients with a cord lipoma was 35 years. The tumors in the sarcoma patients were 10 and 13 cm in greatest dimension compared with a mean size of 5.5 cm (range, 1–8 cm) for the cord lipomas. One of the liposarcomas had radiographic (CT scan) evidence of extension from a retroperitoneal lesion; the other appeared to be confined to the groin. At exploration, the apparent retroperitoneal extension was not confirmed; the lesion was found to consist of inflammation and scar formation deep to the inguinal ring from a prior inguinal hernia repair.

Both cases of inguinal liposarcoma presented as nonreducible ("incarcerated") soft-tissue masses in the inguinal canal extending well into the scrotum. One of the patients was status post resection of a soft-tissue mass from the ipsilateral scrotum 18 years earlier. He had been told that the lesion was benign. Despite physical evidence of incarceration, neither patient had gastrointestinal complaints and both lesions were painless. Both lesions had been present for several years (3–7 years) and were noted to be slowly enlarging. The ipsilateral testicle was readily palpable in both cases. Based on the absence of clinical symptoms, both patients had delayed seeking medical attention for several years.

The first patient, who had had prior scrotal surgery, underwent an incomplete resection at his initial operation. A reexploration was accomplished with completion of the resection, partial scrotal resection, and orchiectomy since the lesion extended down into the scrotum and scarring from the prior operation prevented development of a tissue plane between the tumor and the testicle. The testicle was uninvolved on histologic examination.

The second patient, who had had a recent hernia operation, and who had no evidence of retroperitoneal involvement on CT, underwent exploration and complete resection without orchiectomy.

Histologic Features of Inguinal Liposarcomas

Similar histologic features were identified in the two cases and these were characteristic of well-differentiated liposarcomas. Both showed enlarged hyperchromatic nuclei embedded in otherwise mature-appearing adipose tissue. The atypical nuclei tended to be present in connective tissue septa, a typical finding in such tumors, and one of the cases demonstrated a solid sclerotic nodule that lacked diagnostic features of dedifferentiation (Figs. 1 and 2).

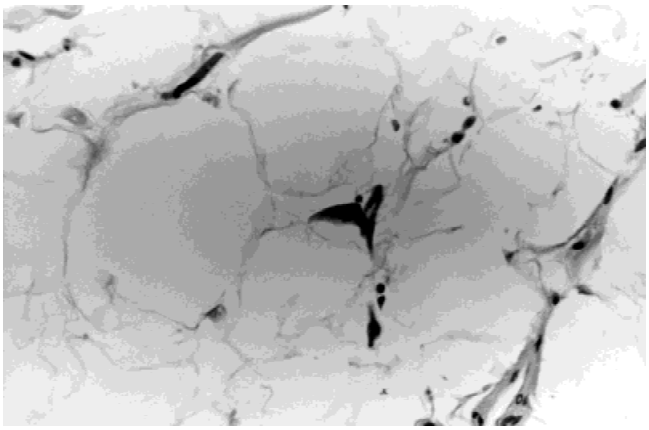

Fig. 1. Well-differentiated liposarcoma. Note the enlarged hyperchromatic cell embedded in a backdrop of mature-appearing adipose tissue. Lipoblasts are not required for a histologic diagnosis of these tumors in clearly lipogenic neoplasms such as this one.

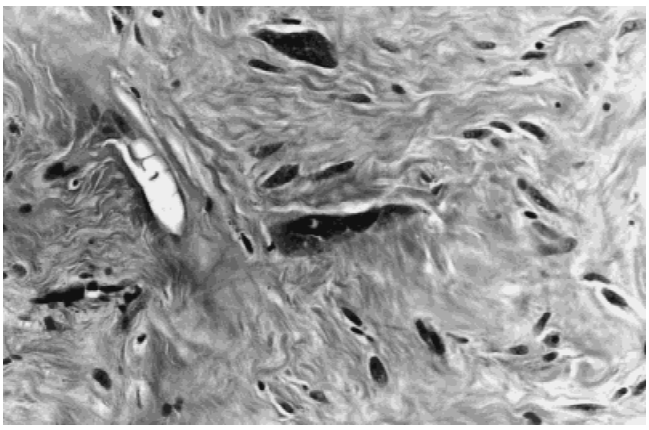

Fig. 2. This sclerotic nodule contained enlarged hyperchromatic cells as well. Such cells frequently course along connective tissue septa in well-differentiated liposarcomas.

Follow-Up Information

Following complete resection, both patients have been followed for 12 months without recurrence. The first patient, who underwent orchiectomy and partial scrotal resection, also received adjuvant postoperative radiation therapy, whereas the other patient received no adjuvant treatment. Follow-up has been by physical examination and CT scan of the pelvis and retroperitoneum. These patients will require long-term follow-up since these lesions may recur after a long latency.

DISCUSSION

Incidental liposarcomas identified during hernia operations are rare (<0.1% at our institution), although this general anatomic site was one of the common unusual sites for liposarcomas in a review of cases accessioned to the Armed Forces Institute of Pathology (AFIP), comprising 130 of 1,067 total cases in archives [1]. In the AFIP material, the inguinal region ranks third, behind the thigh and retroperitoneum. However, this information must be evaluated in the context of the overall incidence of sarcomas. For example, in a large Swedish series, the annual clinical incidence of benign soft-tissue tumors was approximately 3,500 per 1 million patients, whereas the corresponding figure for sarcomas was 18 per 1 million, making benign tumors about 200 times more common than sarcomas [2]. Generally, inguinal liposarcomas are encountered infrequently in practice and they are encountered in the literature principally as case reports [3–10] and as components of larger series of cases. For example, fourteen such cases were a component of 1 series of 92 well-differentiated liposarcomas reported by Weiss and Rao [11].

In the present two cases, both tumors were well-differentiated liposarcomas without evidence of dedifferentiation, in keeping with most reported liposarcomas in this site, although myxoid liposarcomas have been reported in several cases and are best illustrated in one reported by Bauer et al. [3]. Schwartz et al. [8] have recently reported a series of such cases and summarized earlier literature. All six cases reported by Schwartz et al. [8] presented as well-differentiated liposarcomas, although three underwent dedifferentiation and two of the patients in their series had retroperitoneal disease or metastases. In the Weiss and Rao series [11], 11 of 14 groin liposarcomas recurred locally, 4 dedifferentiated, and 2 caused the death of the patient.

Well-differentiated liposarcomas generally present in the deep soft tissues of the extremities, trunk, retroperitoneum, and, as discussed here, the spermatic cord. Microscopic patterns vary, but most cases can be classified as lipoma-like, sclerosing, or inflammatory types. These subtypes have no bearing on prognosis but are a good construct to remind pathologists of the morphologic range of well-differentiated liposarcoma. The surgeon and pathologist must be aware that any large lipomatous neoplasm of the retroperitoneum, mediastinum, or spermatic cord is a liposarcoma until proven otherwise. The World Health Organization [12] defines well-differentiated liposarcoma as “a tumour composed of mature fat cells, occasional atypical hyperchromatic cells and lipoblasts.” These tumors are known to be prone to local recurrence but not to metastases. When such tumors present in the subcutaneous tissues, they are frequently referred to as “atypical lipomas” [13,14]. However, since the inguinal/spermatic cord region communicates directly with the retroperitoneum, all such tumors presenting in this region are regarded as deep and would hence be classified as well-differentiated liposarcomas by virtue of their location. Although most well-differentiated liposarcomas remain histologically bland, a small percentage (about 10%) may undergo histologic progression (dedifferentiation) to a higher-grade lesion. This phenomenon was first described in the setting of low-grade chondrosarcomas that underwent such a progression.

The classic concept of dedifferentiated liposarcoma consists of low-grade liposarcoma juxtaposed to high-grade sarcoma indistinguishable from malignant fibrous histiocytoma. This phenomenon is most likely to occur in bulky retroperitoneal tumors but has been reported in extremity lesions and even in subcutaneous ones [11,15]. Treatment of retroperitoneal lesions is en bloc removal, including adjacent organs and major vessels (retroperitoneal exenteration). In the absence of dedifferentiation and metastases, death in well-differentiated liposarcomas occurs by gastrointestinal/urinary obstruction. Treatment of extremity lesions usually involves complete excision in a function-preserving fashion as these sites are readily monitored and amenable to reexcision in the event of local recurrence. This approach is justifiable since liposarcomas of the extremities rarely metastasize. Generally, the presence of dedifferentiation can be suggested on radiographic studies.

Although the original concept of dedifferentiation as proposed by Evans [14] included specific criteria and required a high-grade component with at least 5 mitoses/10 high-power fields, this concept has evolved over time. Recent studies suggest that even low-grade dedifferentiation, producing a pattern reminiscent of low-grade fibrosarcoma, and small amounts of dedifferentiation still carry the poor prognosis associated with high-grade extensive dedifferentiation [16,17], as these low-grade forms merely reflect an early stage of the transformation to more aggressive neoplasms. The 155 cases of dedifferentiated liposarcoma analyzed by Henricks et al. [17] included 13 spermatic cord/scrotal examples. Follow-up information was available on 11 of these patients; there were 2 recurrences, 2 metastases, and 2 tumor-related deaths.

Most authors have recommended complete wide excision of well-differentiated spermatic cord liposarcomas. This entails exploration high into the internal ring, possible division and resection of the cord and spermatic vessels, and possible orchiectomy. Resection of the hemiscrotum is not necessary if the scrotal skin is uninvolved or has not been violated by prior surgery.

In recent years, economic pressures have been exerted to streamline the number of specimens evaluated histologically. For example, in 1980, Roslyn et al. [7] advocated reviewing histology on all tissue removed at her-

niorrhaphy as it "offers a unique opportunity for peritoneal biopsy that should not be overlooked" after noting the detection of 3 cases of occult malignancy among 1,200 herniorrhaphy specimens (2 adenocarcinomas and 1 liposarcoma). However, by 1986, Kassan et al. [5], who discovered 4 neoplasms in 1,020 hernia repair operations, stated that the aggregate charge of $30,528.00 incurred from all the specimens would translate to a national savings of $18,000,000 by omission of pathologic evaluation of herniorrhaphy specimens. These authors noted that in the four cases where neoplasms were detected, the tissues appeared abnormal in three, suggesting that histologic evaluation was best reserved for cases in which the tissue appeared abnormal at operation. In the present cases, both lesions appeared abnormal to the submitting surgeon. In general, we would suggest evaluation of most excised tissue since the cost of this step is not a major percentage of the overall cost of the patient's care. However, if, as some authors have suggested, efforts to contain costs are implemented and histologic review of herniorrhaphy tissue is deemed generally unrewarding, large (>10 cm) fatty masses and any other unexpected nodules or apparently thickened areas of peritoneal tissue from this area should still be sampled.

Discovery of a large, nonreducible soft-tissue mass at exploration for an apparent hernia presents a challenge to the operating surgeon. Complete surgical extirpation offers the best hope for cure. Incomplete resection may result in further surgery and may increase the risk of local recurrence. Preoperative suspicion of the diagnosis may allow more thorough dissection and submission of tissue to assess margins (although the separation of mature adipose tissue from well-differentiated liposarcoma is difficult at best on frozen sections) and even orchiectomy if necessary.

A high index of suspicion for this lesion may allow definitive surgical care at the first operation. Planned complete resection will result in a low rate of local recurrence.

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